International Journal of Surgery Case Reports 7 (2015) 79–81

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Recurrent iliac hydatidosis: A case report

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ARTICLE INFO

Article history:
Received 4 December 2013
Received in revised form 9 April 2014
Accepted 23 July 2014
Available online 11 December 2014

Keywords:
Hydatid disease
Bone
Recurrence

ABSTRACT

INTRODUCTION: Hydatid cysts may occur in any area of the body, but they usually localize to the liver and the lungs. Primary localization in bone is not common.
PRESENTATION OF CASE: The authors report the case of multifocal hydatid disease appeared 3 years after the surgical treatment of a cyst of the hip. The patient presented with cough and chest pain of 2 months duration. Only the lung localization was symptomatic. The thoracic echography, the abdominal and chest scan allowed the diagnosis.
DISCUSSION: Hydatid recurrence remain frequent, whatever is the nature of the treatment, surgical or chemical.
CONCLUSION: The premature detection of recurrence is of great importance.

1. Introduction

Hydatidosis is a parasitic disease caused by *Echinococcus granulosus.* The bone infection is rare. About 50% of the cases of bone hydatid disease affect the spine; the second most frequent location is the pelvis (21%). Hydatid bone disease is often asymptomatic, and is therefore usually diagnosed at an advanced stage. It is not easy to eradicate and may be impossible to cure.

We report an unusual case of a recurrent iliac hydatid cyst, despite surgical procedure which was performed in this case.

2. Presentation of case

It was a rural 55-year-old woman. She had previous medical history of iliac hydatid cyst in 6 years before (2006). At this time, she presented with painful lumbar mass and pelvic CT scan showed expansible lytic lesions with cortical rupture of the left iliac wing, with multilobulated endo and exo-pelvic collection measuring 18 cm * 10 cm * 15 cm (Fig. 1). She underwent surgery; partial iliac bone resection and endo and exo-pelvic cyst curettage were performed. Histological examination showed typical bone hydatid cyst. It revealed typical hydatid membranes and fibrohyaline wall, associated to florid foreign body giant cell inflammatory reaction. Note that, Chest X-ray before surgery was normal. Unfortunately, our patient did not receive complementary chemotherapy for its pelvic echinococcosis.

In 2012, she was admitted in our department with cough and chest pain of 2 months duration. The clinical examination was poor; it only found fever at 38.6 °C and left lumber sensibility. Chest X-ray showed round culminal opacity with hydro-aeric level (Fig. 2). The abnormal findings on laboratory investigations were: Wight blood cells = 21,000, C-reactive protein = 241 mg/l, Erythrocyte Sedimentation Rate = 59.

According to past history of hydatid disease, the diagnosis of complicated hydatid cyst of the lung was supposed. Additional exams were requested in order to confirm that, and to search other hydatid locations. Abdomen and pelvic sonography revealed liver hydatid cyst type V in the 4 segments, associated to peritoneal and pelvic hydatidosis of the left iliac pit. Body scan confirmed the liver and lung involvement and showed multiple lytic lesions in the left iliac wing and revealing iliac hydatid disease extending to adjacent soft tissues (Figs. 3 and 4). This bone hydatidosis was associated to left iliac deep venous thrombosis, extending to inferior vena cava and complicated by pulmonary embolism.

In conclusion, our patient was presenting multifocal hydatid disease, associating an infected lung hydatid cyst, a liver hydatid cyst type V and a recurrent bone hydatid disease, with iliac deep venous thrombosis, extending to inferior vena cava and pulmonary embolism. She benefited for 6 weeks of antibiotherapy using cefotaxim, aminosid and metronidazol and 6 months of anticoagulation therapy. She has also undergone a surgical extirpation of the lung cyst followed by an ischio-femoral arthrodesis with cystic drainage (it was a palliative treatment), associated to chemotherapy using albendazole administer in the presurgical setting (400 mg orally twice a day with meals). Fig. 5 showed the result of pelvic surgery.

Outcome was favourable but patient presents pelvic pain in walking and no new lesion was, however, seen in chest X-ray at 6 months follow-up.

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http://dx.doi.org/10.1016/j.ijscr.2014.07.018

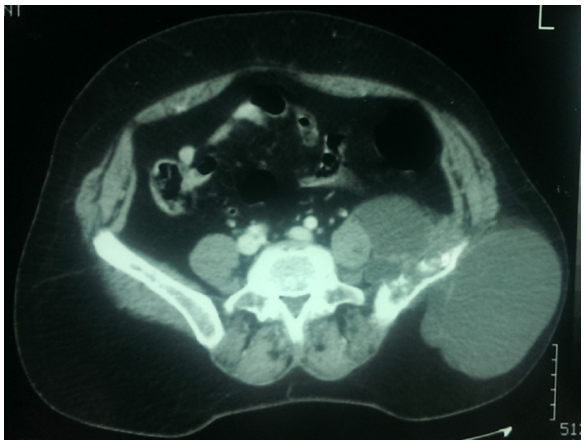


Fig. 1. Pelvis CT scan shows expansible lytic lesions with cortical rupture of the left iliac wing, with multilobulated endo and exo-pelvic collection measuring 18 cm * 10 cm * 15 cm.

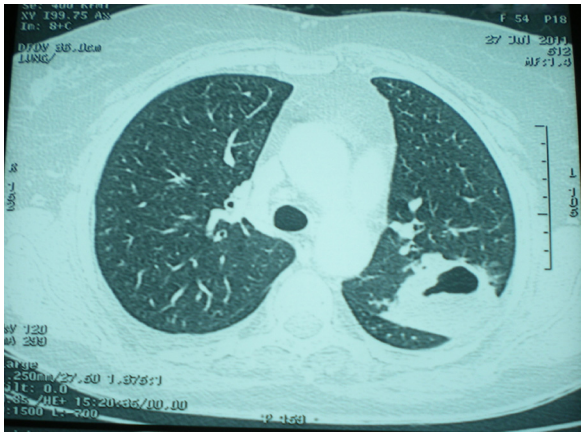


Fig. 2. Chest CT scan shows culminal cavity partially filled, with thickened wall.

3. Discussion

Hydatidosis is a parasitic disease caused by *E. granulosus* by the development of the parasite's larval in man.[1,2] All parts of the body may be involved, but the liver and lungs are the main locations. The bone infection is rare, accounting for 0.5 to 2.5% of all hydatid lesions.[2,3] The vertebrae, ilium, long bones, skull and ribs are most frequently affected. It usually occurs in vascularized areas. Skeletal infestation of *E. granulosus* cyst occurs by haematogenous seeding.[4]

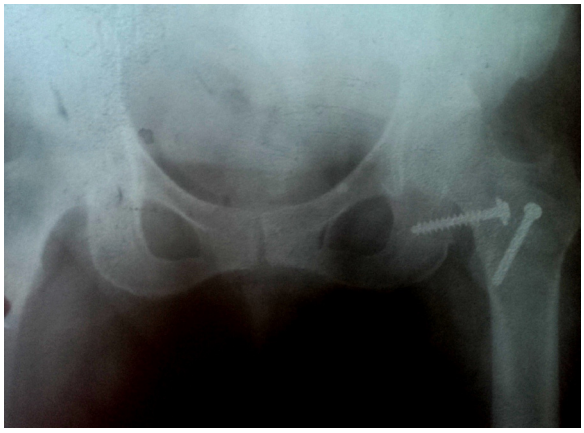


Fig. 5. Pelvis X-ray shows the ischio-femoral arthrodesis.

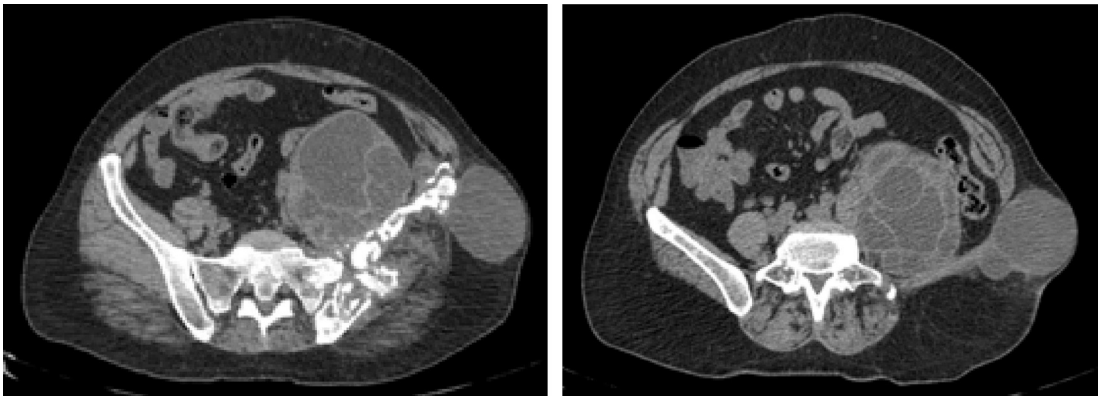
The larval form reaches the bone, penetrates the spongy tissue and grows in the direction of least resistance, infiltrating and damaging the tissue like a tumour.[5] But rigid structure of the bone does not allow the cyst to grow rapidly. This is why it is a very slowly progressive disease and patients are usually asymptomatic and present at an advanced stage of the disease, when lesions have become extensive. There is no delimitation between healthy and pathologic tissues.[4] As no adventitia is formed around the cyst by the host, daughter cysts can spread to adjacent bones. This can explain why bone hydatid disease is polycystic in contrast to other non-osseous locations.[4,6]

The radiological signs include lucent osseous lesions associated with expansion of the bone and thinning of the cortex. In patients with these signs, soft tissue calcification is highly suggestive of hydatid disease.[6] MR Images and computed tomography are valuable in delineating the extent of bone and soft tissue abnormalities.[7]

So in endemic regions, because of the diversity of its presentation, hydatid disease should always be in the differential diagnosis list of osteolytic lesions or any growing destructive mass.

The diagnosis is often made on the basis of the characteristic radiographic appearance of the lesions. Eosinophilia is seen in only 25% of cases. Serological tests are often negative when hepatic and lung disease are absent. Although histological study is diagnostic, needle biopsy may lead to dissemination of the infection; it can be responsible for anaphylactoid reactions.[6]

The treatment and prognosis of bone hydatid disease resemble those of a malignant tumor.[4] Surgery is the treatment of choice with or without chemotherapy. Radical and wide resection of the



Figs. 3 and 4. Abdomino-pelvis CT scan show osteolysis of the left iliac wing with large multilocated retroperitoneal hydatid cyst of 16 cm with invasion of the left psoas muscle and hydatid cyst of subcutaneous fat regarding left iliac crest of 7 cm.

involved bone with the surrounding soft tissue is recommended except in areas such as the pelvis, which is technically impossible.[4,6] Curettage and bone graft is another procedure but with local recurrence rate of 70–80%.[8] Adjuvant medical therapy using albendazole or mebendazole may be added to surgery when complete excision of the cyst is not possible or doubtful[5,9,10]; or used like isolated therapy when surgical treatment is not possible because of extensive involvement.[9] It can control the disease locally, prevent recurrence and avoid systematic spread. A treatment period of 2 years may be necessary, although sometimes lifelong treatment is recommended.

The prognosis is poor when bone is involved, even in patients who undergo extensive medical and surgical treatment,[5] such as is the case of our patient, in whom the bone pelvic hydatidosis had recurred despite surgical treatment.

4. Conclusion

Hydatid bone disease is often asymptomatic, and is therefore usually diagnosed at an advanced stage, it is not easy to eradicate and may be impossible to cure. Its prognosis is poor. Surgery is the treatment of choice. Radical resection of the involved segment, with or without chemotherapy, is recommended except in areas such as the pelvis, which is technically impossible, justifying the recurrence frequency even in patients who undergo extensive medical and surgical treatment.

Conflict of interest

None.

Funding

None.

Ethical approval

Authors confirm that consent has been obtained.

Author contributions

Ben Amar j, Zaibi H: data collection, data analysis, and writing; Bouacha H, Aouina H, Dahari H: contributors.

Key learning points

- Hydatid bone disease is often asymptomatic, and is therefore usually diagnosed at an advanced stage. It is not easy to eradicate and may be impossible to cure.

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